

## Case Report

# A Case of Rhinosporidiosis and Concomitant Inverted Papilloma - A Review of the Literature

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- Rhinology
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## Abstract

Rhinosporidiosis is a chronic granulomatous disease of mucous membranes and is endemic in Southern India and Sri Lanka, though there are infrequent reports of the disease elsewhere in the world with *Rhinosporidium seeberi* identified as aetiological agent. Sino nasal Inverted papilloma constitutes only 0.5-4% of all nasal tumours and may be associated with human papilloma virus, chronic inflammation, allergy and occupational exposures. We present a case in which these two rare pathologies presented synchronously in a patient presenting with unilateral nasal obstruction secondary to nasal mass. This is the first case report of rhinosporidium in a white Caucasian patient without any history of travelling to endemic area. Concomitant presence of inverted papilloma makes it interesting and supports the inflammatory theory as a cause for inverted papilloma.

## ABBREVIATIONS

PAS: Periodic Acid-schiff; HPV: Human Papilloma Virus; CT: Computed Tomography; H&E: Haematoxylin and Eosin

## INTRODUCTION

Rhinosporidiosis is a chronic granulomatous disease of mucous membranes and is endemic in Southern India and Sri Lanka, although there are infrequent reports of the disease elsewhere in the world, including the American subcontinent. *Rhinosporidium seeberi* has been identified as the aetiological agent. Having previously been considered a fungal disease, recent 18S ribosomal ribonucleic acid gene analysis has placed it in a group of aquatic parasites [1-3].

The condition has a male preponderance with a ratio of 4:1 and targets individuals between the second and fifth decades [1]. The natural habitat of the rhinosporidium organism is ground water in ponds, lakes or soil and it is thought to be contracted by local inoculation in the setting of water activities, with infection of the nose and nasopharynx documented to occur in 85% of cases. It can also affect the lips, palate, uvula, ear, conjunctiva, larynx and trachea [1].

Histologically the mature endospores are thick walled and stain positively with periodic acid-schiff (PAS) or Grocott's silver stain. Currently diagnostic status rests mainly on the morphological and staining characteristics of different stages of the *rhinosporidium* [4].

Rhinosporidial growths are of variable duration (8 days -35 years) although in ocular rhinosporidiosis associated with a preceding event like trauma the incubation period may be less than this [4]. Clinically, rhinosporidiosis appears as mass which is painless, slow growing, papillomatous and/or polypoidal with the tissue often soft, friable and erythematous due to increased vascularity with a definitive diagnosis based upon histology [4]. Treatment is by surgical excision when accessible by electro cauterization of the base of the excised site although cryosurgery can also be used. Systemic therapy with dapsone (diaminodiphenyl sulfone) serves as an alternative treatment option [4].

Sino nasal Inverted papilloma initially described by Ward in 1854 and Billroth in 1855 constitutes only 0.5-4% of all nasal tumours [5]. The peak presentation is in the sixth decade, with a male to female ratio of between 3:1 and 10:1 and it is most commonly found on the lateral nasal walls and in the sinuses. Inverted papilloma have a propensity for recurrence, local aggressiveness, transformation to carcinoma and high risk of synchronous or metachronous malignancy [5]. It has been hypothesised that human papilloma virus (HPV) may be associated with the development of inverted papillomas; particularly HPV 11 [6]. Other suggested aetiological factors include chronic inflammation, allergy and occupational exposures. Even after adequate treatment of an inverted papilloma, it has a high tendency to recur in up to 30 - 60 % of cases [2].

We present the first documented case of these two rare

pathologies presenting synchronously in a patient presenting with unilateral nasal obstruction.

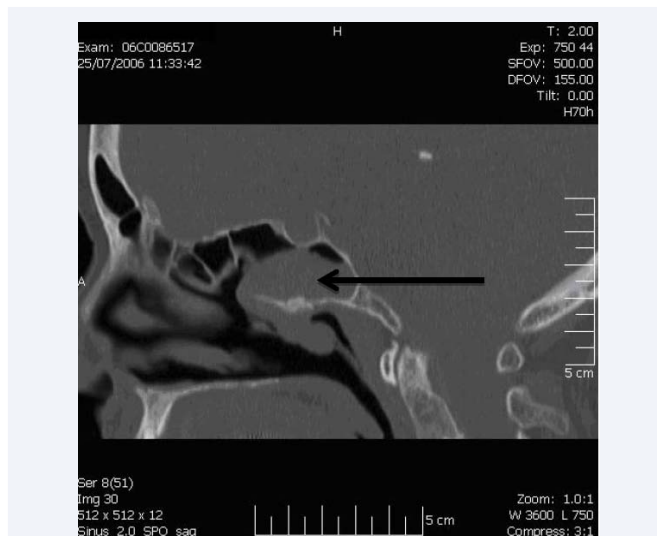
**CASE PRESENTATION**

A 78 year old woman presented with a five year history of right sided nasal obstruction, previously treated with a topical intra-nasal steroid spray. There was no associated epistaxis, anosmia, facial pain or headaches and she had an unremarkable past medical history. There was no travel history to endemic areas.

Detailed ENT examination demonstrated a polypoidal mass arising from sphenothmoidal area in the right nasal cavity (Figure 1). The left nasal cavity was normal and there was no sinus tenderness or lymphadenopathy. A computed tomography (CT) scan of the nose and paranasal sinuses was arranged pre-operatively (Figure 2&3). The scan showed soft tissue and mucosal thickening in the nose, nasopharynx and the right sphenoid sinus with features of chronic osteitis and the possibility of neoplasm was suggested.

At surgery a large polyp and mucopus originating from the right sphenoid sinus was removed and sent for histology (Figure 4). The patient was discharged later that day and made an uneventful recovery.

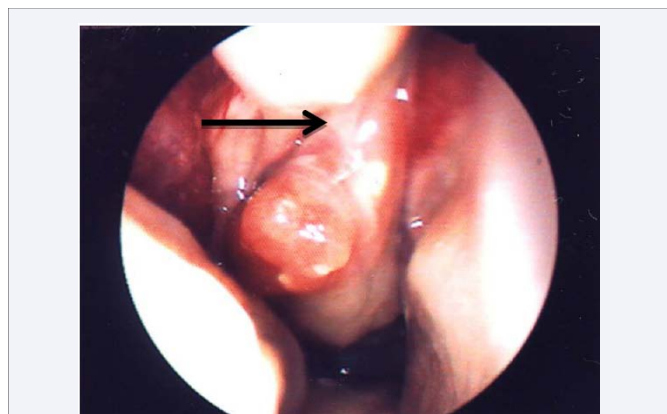
Histology demonstrated two distinct histological entities. On routine haematoxylin and eosin (H&E) staining there was evidence of invagination of the surface epithelium into the underlying stroma, with patterns of mitosis in the basal layer (Figure5). There were cysts present containing spores with PAS positive walls and electron dense bodies (spherules) in sporangium (Figure 6), a silver stain was applied (Figure 7) and showed that some of these spores were lying freely in the nasal tissue. These two findings are consistent with two pathologies occurring synchronously: rhinosporidiosis with concomitant inverted papilloma of the paranasal sinuses. At 3, 6 and 12month follow-up the right sphenoidotomy was clean and healthy with no signs of recurrence of disease.



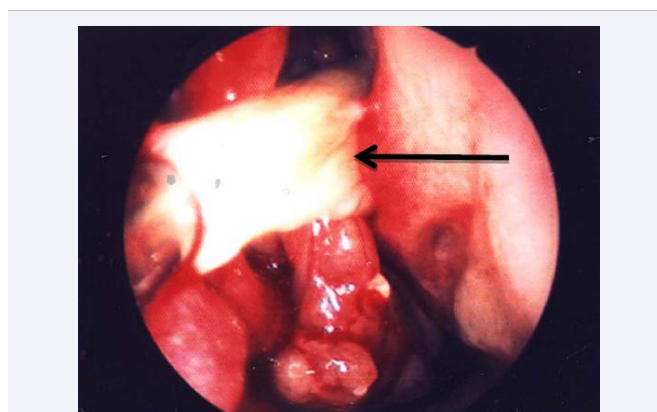
**Figure 2** Computed tomography scan, a sagittal view of right nasal mass arising from the sphenoid sinus.



**Figure 3** Computed tomography scan, an axial view of right nasal mass arising from the sphenoid sinus.



**Figure 1** Polypoidal mass arising from sphenothmoidal area in the right nasal cavity.



**Figure 4** Mucopularising from the right sphenoid sinus.

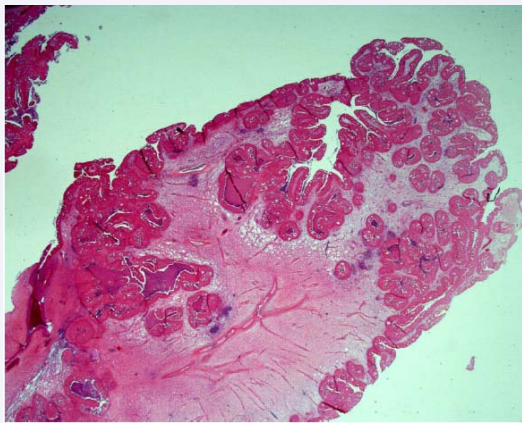


Figure 5 H&E x10. Inverted papilloma.

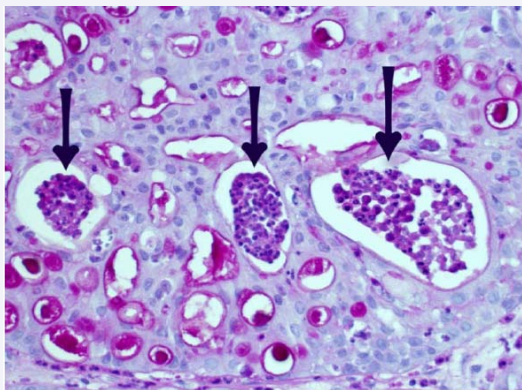


Figure 6 Positive PAS x 400, staining demonstrating presence of rhinosporidium.

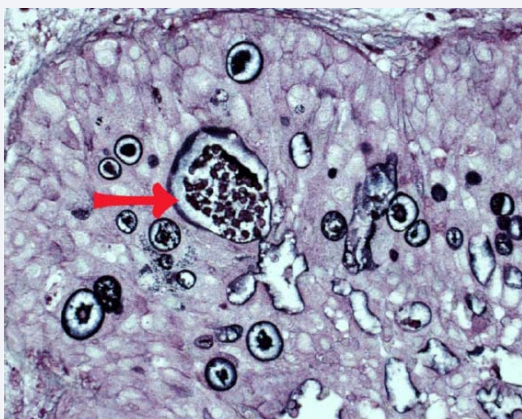


Figure 7 Positive Grocott's silver x 400. Identifying sporangium with endospores.

## DISCUSSION

Historically the exact phylogenetic affinities of *Rhinosporidium seeberi* have been controversial. Seeber's thesis described rhinosporidiosis in 1900, considered the sporangium of this enigmatic organism to be a sporozoan allied to the coccidian

[10]. Ashworth concluded the organism is closer to lower fungi (phycomycetes) [6]. Large and subepithelial sporangia are visible as pale spots, the size of pin head on the vascular polyps which then have a characteristic of strawberries. The endospores represent the asexual spores of the organism. These are embedded in mucoid material which stains positively with alcian blue and mucicarmine stains the mucoid material which is a mucopolysaccharide inhibits phagocytosis of the organism in the host tissue and prevents penetration of drugs [4].

The term inverted papilloma describes the histological appearance of the epithelium as inverting into the stroma with a distinct and intact basement membrane that separates and defines the epithelial component from the underlying connective tissue stroma [7]. Inverted papilloma is associated with tendency to recur, destructive potential, association with nasal polyps and propensity to be associated with malignancy. HPV 11 with chronic inflammation has been described as important factors in the development of these tumours [8]. Common presentations are nasal blockage, congestion, epistaxis, nasal discharge and recurrent sinusitis. Chronic osteitis and hyperostosis have been reported to be important preoperative CT scan findings [9]. 11% of recurrent papillomas develop into metachronous malignancy with the mean time for the development of carcinoma was found to be 52 months [10].

Management is surgical and based on the preoperative CT scan the preliminary extent of the tumour should be assessed. To gain adequate clearance of the disease representative biopsy with frozen section can be helpful. Naso-ethmoid tumours have a better prognosis than maxillary, frontal and sphenoid [10]. 2 year follow up is essential minimum requirement for preliminary report of treatment results and life-long follow up because of the possibility of malignant potential and chances of recurrence [10].

This is the first case report of rhinosporidium in a white Caucasian patient without any history of travelling to endemic area. Concomitant presence of inverted papilloma makes it interesting and supports the inflammatory theory as a cause for inverted papilloma.

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